Lymphoplasmacytic Sclerosing Pancreatitis Finding After Laparoscopic Nissen Fundoplication Mimicking Malignancy: Case Report

Laparoskopik Nissen Fundoplikasyonu Sonrası Saptanan Maligniteyi Taklit Eden Lenfoplazmositik Sklerozan Pankreatit

Mehmet Tolga KAFADAR,[a]
Metin YALAZA,[b]
Önder SURGİT[a]

[a]Department of General Surgery, Turgut Özal University Faculty of Medicine, Ankara
[b]Clinic of General Surgery (Surgical Oncology), Konya Training and Research Hospital, Konya

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Yazışma Adresi/*Correspondence:*
Mehmet Tolga KAFADAR
Turgut Özal University Faculty of Medicine, Department of General Surgery, Ankara, TÜRKİYE/TURKEY
drtolgakafadar@hotmail.com

ABSTRACT Also known as autoimmune pancreatitis, duct destructive chronic pancreatitis, sclerosing pancreatitis, or primary inflammatory pancreatitis, lymphoplasmacytic sclerosing pancreatitis (LPSP) is a benign disease of pancreas, although its clinical presentation may be similar to that of pancreatic ductal adenocarcinoma. A 65-year-old male with a past medical history of laparoscopic Nissen fundoplication with mesh hiatoplasty, initially presented to our clinic with intermittent epigastric pain. Imaging studies showed a mass concerning the pancreatic body and tail, with involvement of the coeliac truncus, common hepatic, splenic, superior mesenteric arteries and superior mesenteric vein. A preliminary diagnosis of fibrosis and lymphoplasmacytic infiltration was based on tru-cut biopsy. The patient underwent a laparoscopic pancreatic biopsy. The final pathological diagnosis was LPSP. To the best of our knowledge, this is the first reported case of a LPSP developing after a laparoscopic Nissen fundoplication.

Key Words: Laparoscopy; fundoplication; pancreatitis

ÖZET Lenfoplazmositik sklerozan pankreatit (LPSP), klinik olarak pankreas duktal adenokarsinomunu taklit edebilen benign bir hastalıktır. Otoimmün pankreatit, destruktif kronik pankreatit, sklerozan pankreatit ve primer inflamatuar pankreatit olarak da bilinir. Mesh hiatoplasti ile Laparoskopik Nissen fundoplikasyonu öyküsü bulunan 65 yaşında erkek hasta kliniğimize aralıklı epigastrik ağrı şikâyeti ile başvurdu. Görüntüleme tetkiklerinde pankreas korpus ve kuyruk kısmında, çölyak trunkus, common hepatik, splenik, superior mesenterik arteri ve superior mesenterik veni de içine alan kitle saptandı. Tru-cut biyopsi ile fibrozis ve lenfoplazmositik infiltrasyon ön tanısı konuldu. Hastaya laparoskopik pankreas biyopsisi yapıldı. Patolojik tanısı LPSP olarak rapor edildi. Bilgilerimiz dahilinde, bu olgu Laparoskopik Nissen fundoplikasyonu sonrası gelişen, bildirilmiş ilk LPSP vakasıdır.

Anahtar Kelimeler: Laparoskopi; fundoplikasyon; pankreatit

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Lymphoplasmacytic sclerosing pancreatitis (LPSP) is an autoimmune disorder, clinically mimicking pancreatic cancer and must be resected if malignant features are present. The diagnosis is based on the diagnostic criteria of autoimmune pancreatitis proposed by the Japan Pancreas Society in 2002, that were modified in 2006.[1,2] Elevated levels of serum IgG4 suggest a diagnosis of LPSP preoperatively and may differentiate it from pancreatic cancer.[3] Herein, we present LPSP finding after laparoscopic Nissen fundoplication mimicking malignancy.

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CASE REPORT

A 65-year-old male presented with a 5-year history of gastroesophageal reflux disease. The patient opted for a long-lasting surgical solution, rather than relying on life-long proton pump inhibitor treatment. The endoscopic examination revealed a hiatal hernia of 3 to 4 cm in diameter, as well as reflux esophagitis of moderate severity. Abdomen ultrasonography was normal. Reflux disease was confirmed by esophageal manometry and pH studies. A short 360-degree Wrap bound with Ethibond that involved the esophageal wall was used to carry out a laparoscopic Nissen fundoplication. The patient's hiatus was also repaired with plication of the crura and mesh hiatoplasty was performed. Both vagus nerves were identified and left intact. He had an uneventful recovery and was discharged on the first postoperative day.

Six months later, at an outpatient office visit, he complained of persistent, mild mid-epigastric pain radiating to the back and denied any alcohol or medication use, history of infection or trauma. He had no history of gallstone and autoimmune disorders. Abdomen and chest physical examinations were normal. Abdomen ultrasonography showed a pancreatic mass localized at the pancreatic body and tail. Computed tomography (CT) of the abdomen demonstrated diffuse, isohomogenous mass of the pancreas predominantly involving the pancreatic body and tail with encasement of the mesenteric, common hepatic, coeliac, splenic and right renal vessels (Figure 1). Laboratory studies revealed normal serum levels of amylase, lipase, liver function tests, CA19-9 and CEA levels. Moreover, his all infectious work-up were negative.

Given the typical presentation for pancreatic adenocarcinoma (PA) and the tomography findings, a preliminary diagnosis of PA was entertained. Tru-cut biopsy of the pancreas was performed and reported as consistent with fibrosis and lymphoplasmacytic infiltration. Immunglobulin and autoimmune markers were evaluated with the suspicious diagnosis of LPSP. Antinuclear antibody was positively detected. Other autoimmune markers and total IgG, IgG4 levels were all in normal range. The patient underwent laparoscopic pancreatic biopsy. During operation, there was a large, stiff bulky mass in the body and tail of the pancreas with a moderate amount of pancreatic edema. Frozen sections of the specimen were negative for malignancy. The pathological report was chronic pancreatitis secondary to LPSP without evidence of malignancy (Figure 2A-C). The patient had an uneventful postoperative course and was discharged home on day 1. Prednisone 0.5-0.9 mg/kg per day was started and the dose was reduced to 5 mg/wk after 4 weeks. His complaints were significantly improved and follow-up over one year after laparoscopic pancreatic biopsy was uneventful. A control CT could not be taken for our patient since he did not attend routine control visits beyond 1 year after laparoscopic pancreas biopsy sampling.

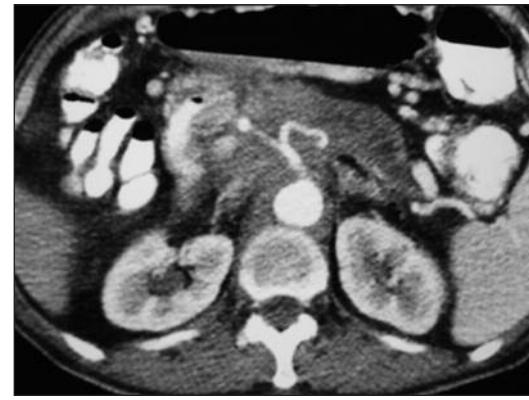

FIGURE1: Image of pancreatic mass with computed tomography.

DISCUSSION

Lymphoplasmacytic sclerosing pancreatitis is a type of chronic pancreatitis of autoimmune origin that is characterized by diffuse fibrosis of the pancreatic tissue and associated with inflammatory bowel disease, Sjögren's syndrome, and some forms of atopy.[3] In our patient, none is present. Another concomitant autoimmune disorder may suggest that LPSP is an autoimmune disorder, but in large series, this is found in no more than 25% of all patients with LPSP. Histopathologically, LPSP has classical and intermediate forms. The former is characterized by pancreatic infiltration by lymphoplasmacytic cells, interstitial fibrosis, periductal inflammation, and

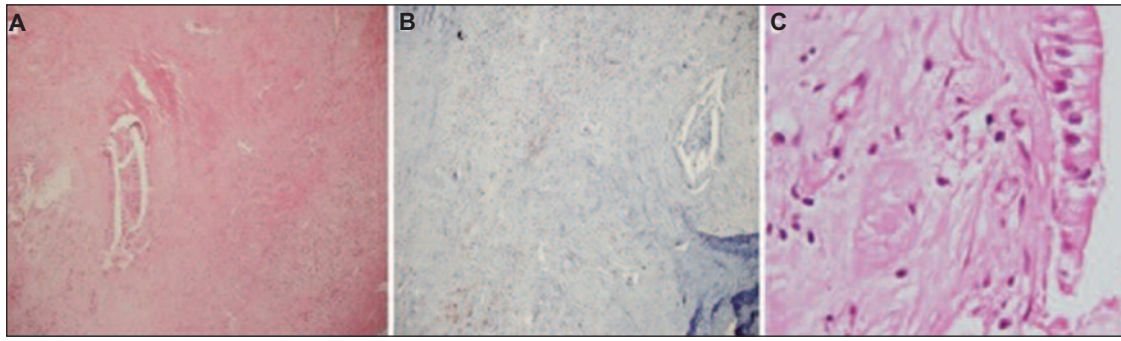

FIGURE 2A: Mononuclear inflammatory cells in the hypocelluler fibrotic connective tissue which surrounds ductus. **B:** Immunohistochemically CD38+ plasma cells between the mononuclear inflammatory cells in the fibrotic area. **C:** Nuclear polarity in the basal membrane of the ductus epithelia and plasmasitoid like mononuclear inflammatory cells in the adjacent connective tissue.

periphlebitis, while the second form is said to be present when at least two of the above histological features are present in a patient.[4] LPSP has a similar clinical course with that of PA in that both conditions reportedly cause cachexia, jaundice, increased CEA and CA 19-9 levels. However, a solitary mass lesion points to PA and the enlargement of the whole gland is more likely to indicate LPSP.[5] An accurate tissue diagnosis is required prior to attempting a surgical procedure or a medical therapy. Effective steroid therapy for LPSP obviates the need for surgical resection. Six to eight weeks of steroid therapy is required to treat LPSP successfully.[6]

In previous studies, it has been shown that LPSP was associated with high serum levels of IgG4 and may be used to distinguish it from pancreatic cancer. Importantly, these levels are not necessarily higher in LPSP, as in our patient.[7] Although IgG4 levels, when elevated, are 95% specific to LPSP, it is a common practice to obtain a histological diagnosis before commencing treatment, especially when immunoglobulin levels are normal.[8] In the case when there are normal IgG4 levels and no surgery is contemplated, a biopsy is usually required to rule in LPSP and exclude PA. It has been frequently reported that LPSP cannot be readily diagnosed by percutaenous biopsy of pancreas, and therefore it is not appropriate to administer a non-interventional treatment when a cancerous lesion cannot be reliably excluded. This is especially true because there is actually an uncertainty in the literature regarding the diagnostic pathological criteria of LPSP.[9] Lymphoplasmacytic sclerosing pancreatitis has been diagnosed on fine needle aspiration cytology and core biopsies in some studies but required open biopsy in others. We diagnosed our patient by obtaining a laparoscopic biopsy of pancreas, the pathological examination of which revealed the classical lymphoplasmacytic infiltration accompanied by sclerosis, periductal inflammation, and phlebitis.

Lymphoplasmacytic sclerosing pancreatitis has a presentation that can mimic cancerous abdominal conditions and can thus be treated with a parallel aggressive approach. It is, however, a necessity to resect an intraabdominal mass lesion that bothers both caregivers and patients to rule out any underlying malignant formation.[10] But in our case surgical resection was impossible because of diffusiveness of the disease. LPSP is medically treated by the administration of steroids (prednisone 0.5-0.9 mg/kg/day or 40 mg/day for 4 weeks), the dose of which is then reduced (5mg per week for another 4 weeks). Steroids usually elicit a favorable response in this condition.[6]

We routinely apply mesh hiatoplasty procedure in laparoscopic Nissen fundoplication operations. We think that mesh hiatoplasty procedure minimizes recurrence risk. In our patient, the mesh that had been placed to the hiatus in the first operation could not be clearly seen in the second operation when the laparoscopic pancreas biopsy was

taken, because of diffuse intraabdominal adhesions and fibrosis. Hence, we are unable to provide any definitive information as to whether mesh had been displaced or removed.

In conclusion, to the best of our knowledge, this is the first reported case of LPSP after laparoscopic Nissen fundoplication and mesh hiatoplasty. We speculate that the fibrosis related to the mesh used for hiatoplasty might have triggered the formation of LPSP. But the manifestation of relation between the mesh and LPSP requires more cases. We wish to alert the physicians to keep in mind LPSP in the differential diagnosis of abdominal pain with pancreatic mass occurred after laparoscopic Nissen fundoplication.

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